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Original

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Utero-ovarian varices and absent inferior vena cava in pregnancy

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Running title

Absent IVC in pregnancy

Abstract

Extensively distended and tortuous vessels on both sides of the uterus are rare incidental findings on transvaginal sonography in early pregnancy. A 31 year-old woman with a history of surgical repair for congenital intestinal stenosis was noted to have utero-ovarian varices on a transvaginal ultrasound examination during her first prenatal visit at six week's gestation. Magnetic resonance imaging revealed dilated ovarian veins along with infrarenal absence of inferior vena cava as well as the absence of external and common iliac veins. Despite concerns regarding spontaneous utero-ovarian vessels rupture, her antenatal course was uneventful. A vaginal delivery was successfully achieved without any postpartum complication. We also demonstrated an intraoperative view of the utero-ovarian vessels during cesarean section in her subsequent pregnancy. IVC absence and pelvic varices can occur in women with a history of neonatal surgery. Although the risk of utero-ovarian varices rupture remains unclear, vaginal delivery may be safely achieved.

Key words Utero-ovarian varices; absent inferior vena cava; magnetic

resonance imaging; mode of delivery; pregnancy

Introduction

The presence of utero-ovarian varices (UOV) in pregnancy raises concerns for potentially catastrophic spontaneous rupture of the vessels, due to mechanical compression of the inferior vena cava and pelvic veins by an enlarged gravid uterus, an increase in intravascular volume, and hormonal factors ⁽¹⁾. Indeed, spontaneous rupture of utero-ovarian vessels was reported in more than 100 cases ⁽²⁾. However, UOV are quite rare, and the rupture of UOV has not been reported in pregnant women. Consequently, it has yet to be determined whether the size of UOV alters as pregnancy advances, and whether continuation of pregnancy and vaginal delivery can be allowed in women with UOV. We herein report a case of large UOV in pregnancy that succeeded in an uneventful vaginal delivery.

Case report

A 31-year-old nulliparous woman with a history of surgical repair for

congenital intestinal stenosis as a 7-day-old neonate was referred to our hospital at 16 weeks' gestation due to bilateral distended and tortuous uterine vessels. Transvaginal ultrasonography demonstrated extensive UOV (6.2 x 6.2 cm) with dilated vessels (up to 2.5 cm in diameter), especially on the right side of the uterine cervix (Figure 1A). Magnetic resonance imaging (MRI) revealed bilateral dilated ovarian veins and the absence of infrarenal inferior vena cava (IVC) and iliac veins. Venous flow from the lower extremities entered the pelvic cavity mainly through the obturator and inguinal canals, forming anastomotic collateral vessels (Figure 1B). The patient and her family were thoroughly informed about the potential risks of foetal and maternal morbidity and mortality from spontaneous rupture of UOV. As they strongly wished to continue the pregnancy, she was closely monitored for the potential risk of UOV rupture. Serial ultrasound and MRI examinations showed no increase in the size of the UOV despite mechanical compression of the pelvic veins by an enlarged gravid uterus, an increase in intravascular volume, and hormonal factors. D-dimer levels did not increase during pregnancy. The patient remained asymptomatic with reassuring foetal status and appropriate foetal growth. A controlled labour induction was conducted at 37 weeks' gestation. She delivered a

2654 g healthy baby vaginally without the Valsalva manoeuvre. The size of the UOV remained unchanged during labour, delivery, and the postpartum period. Low-molecular-weight heparin was administered for venous thromboembolism prophylaxis. The postpartum course was uneventful. One year later, she underwent an emergent cesarean section for preterm breech presentation due to spontaneous onset of labour at 25 weeks' gestation. During the operation, dilated uterine veins were identified between the leaves of the broad ligaments (Figure 1C). Contrast-enhanced computed tomography confirmed the absence of infrarenal IVC and external and common iliac veins (Figure 1D).

Discussion

It is unknown whether pregnancy enhances the risk of spontaneous UOV rupture. Although a single case of spontaneous rupture of collateral vessels was reported in a non-pregnant woman with agenesis of the IVC ⁽³⁾, no report has shown an association between absence of the IVC and spontaneous rupture of UOV during pregnancy. To date, two cases of pregnancy with pelvic varices and absent

infrarenal IVC have been reported, presenting with uncomplicated perinatal course^(4,5). This is the third case of UOV presenting with absent IVC in pregnancy, and each case succeeded in having a vaginal birth without UOV rupture. In the current case, serial evaluation of UOV revealed no alterations in the size of UOV during pregnancy and labor. To date, there is no consensus on the mode of delivery in a rare case of UOV with agenesis of the IVC, due to the lack of a series of similar patients in the literature. However, if the size of the UOV remains unchanged during pregnancy, vaginal delivery may be acceptable at the institution where a multidisciplinary team can be assembled.

The absence of infrarenal IVC is extremely rare in literature, and is not thought to be a congenital abnormality⁽⁶⁾. The embryogenesis of the IVC is well described⁽⁷⁾. IVC develops between the sixth and eighth weeks of embryonic life through the formation, regression, and fusion between three pairs of primitive veins: the postcardinal, subcardinal, and supracardinal veins (in order of appearance). The supra-renal IVC develops from the right subcardinal vein. The renal IVC is formed from the anastomosis between the supracardinal and subcardinal veins. The infrarenal IVC develops from the confluence of the common iliac veins, the caudal

extremity of the right postcardinal vein, the right postcardinal-supracardinal anastomosis, part of the right supracardinal vein, and the right supracardinal-subcardinal anastomosis. Absence of the entire IVC suggests that all three paired venous systems failed to develop properly. In contrast, absence of infrarenal IVC necessitates failure of development of both the postcardinal and supracardinal veins, with preservation of the azygos vein. Since it is difficult to identify a single embryonic event that can lead to isolated absence of infrarenal IVC, the absence of infrarenal IVC is thought to result from intrauterine or perinatal IVC thrombosis, rather than congenital anomalies. This hypothesis is supported by previous reports of agenesis of the infrarenal IVC that is accompanied with thrombosis in a fetus or a newborn^(8,9) or perinatal IVC thrombosis probably due to central venous and cardiac catheterization^(10,11). Hence, it is reasonable that infrarenal IVC is often associated with thrombophilia^(4,5), and the history of surgery or examination in infants that requires central venous and cardiac catheterization^(10,11). In the current case, the patient had neither history of venous thromboembolism nor laboratory data to raise clinical suspicion of inherited thrombophilias. Her neonatal surgery was, therefore, likely to cause both iliac veins and infrarenal IVC

thrombosis, resulting in UOV.

Absent IVC is associated with idiopathic varicose veins and deep venous thrombosis in the lower extremities ^(6-8,11,12). Indeed, absence of IVC is present in about 5% of cases of deep venous thrombosis in young patients ⁽¹³⁾. Moreover, 30% of infants and children who underwent extensive IVC thrombosis will develop post-thrombotic syndrome within 10 years ⁽¹⁴⁾. In the present case, antithrombotic prophylaxis with use of low-molecular-weight heparin was considered during pregnancy, but D-dimer levels did not increase during pregnancy. So, low-molecular-weight heparin was administered for venous thromboembolism prophylaxis only during the postpartum period.

To the best of our knowledge, this is the first report to demonstrate an intraoperative view of the UOV beneath the layers of broad ligaments, which were less apparent than expected. This finding raised the concern that venous return from the lower extremities could be blocked by the ligation of uterine veins without perception in case of hysterectomy. Every pregnant woman can face uncontrolled life-threatening postpartum hemorrhage, and may require hysterectomy unexpectedly. In cases with agenesis of both infrarenal IVC and iliac veins, hysterectomy may

cause severe lower extremity edema. Neonatal surgery has become increasingly common and the incidence of pregnancy with acquired IVC absence will increase. If pelvic varices are incidentally observed by ultrasonography in pregnancy, a careful evaluation should be performed to review the patient's medical history and to exclude the absence of iliac veins and IVC.

In summary, vaginal delivery was successfully achieved without any complication, and the size of UOV remained unchanged during pregnancy, labor, and postpartum. Continuation of pregnancy and vaginal delivery may be allowed in women with UOV despite the risk of UOV rupture during pregnancy and labor.

Disclosure of Interests

The authors report no conflict of interest.

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Figure Legends

Figure 1. Utero-ovarian varices.

(A) Transvaginal ultrasound images of pelvic varices at 16 weeks of gestation.

Multiple dilated and tortuous structures with a tubular appearance are seen in the right side of the uterine cervix. (B) True FISP images of utero-ovarian varices with absent inferior vena cava (IVC). The left image shows dilated ovarian veins (arrowhead) along with the absence of infrarenal IVC. Venous blood from the lower extremities entered the pelvic cavity mainly through the obturator (arrow) and

inguinal (double arrow) canals. (C) Intraoperative view of utero-ovarian varices.

Dilated uterine (arrow) and ovarian (double arrow) vessels are seen beneath the layers of the broad ligaments. (D) Three-dimensional image of utero-ovarian varices with absent IVC created from an abdominal CT scan. Uterine varices (oval) in the right side of the uterus (arrowhead) and the dilated ovarian vein (arrow) are seen at 3 months postpartum.

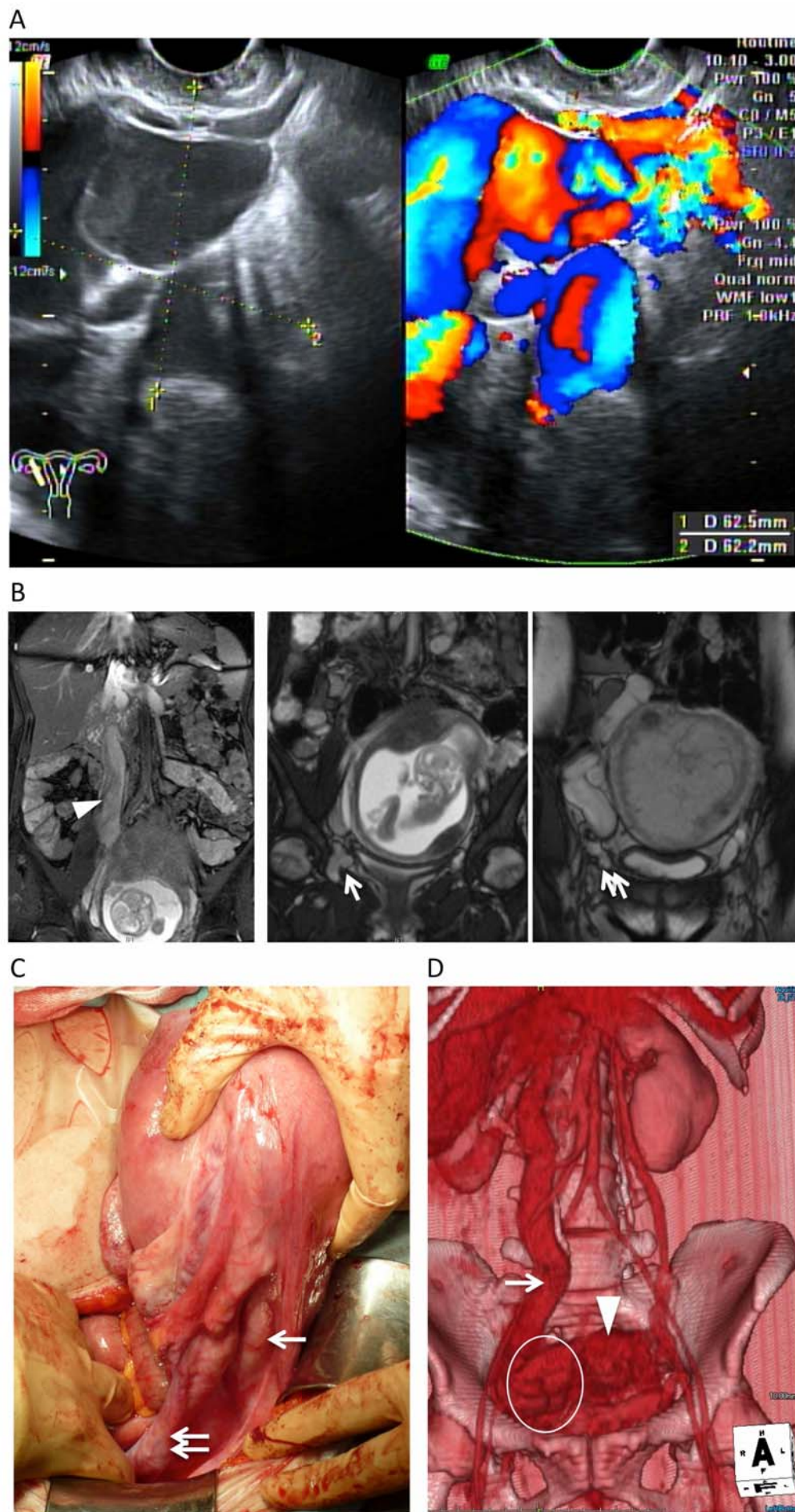


Figure 1